

ACT for Chronic Pain in Neurofibromatosis Type 1: A Randomized Controlled Trial

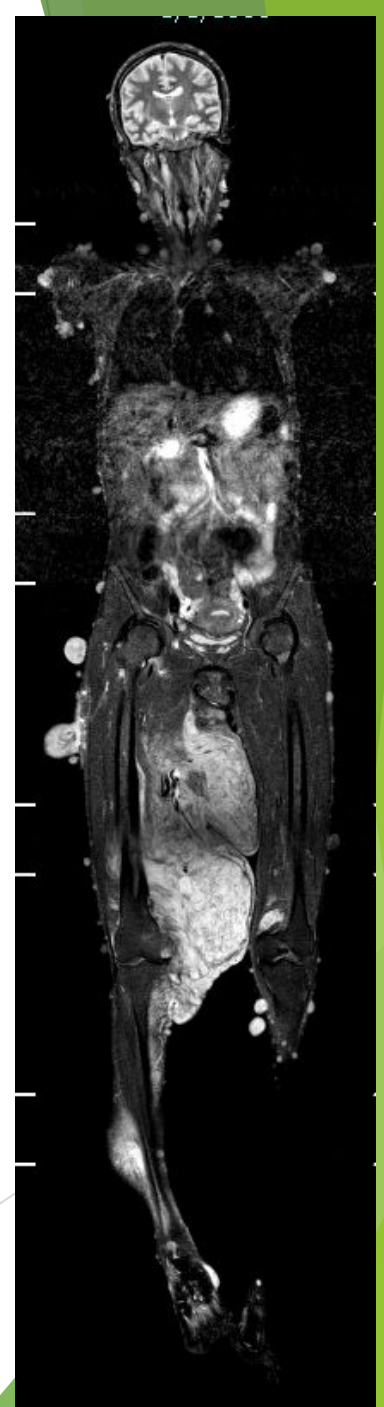
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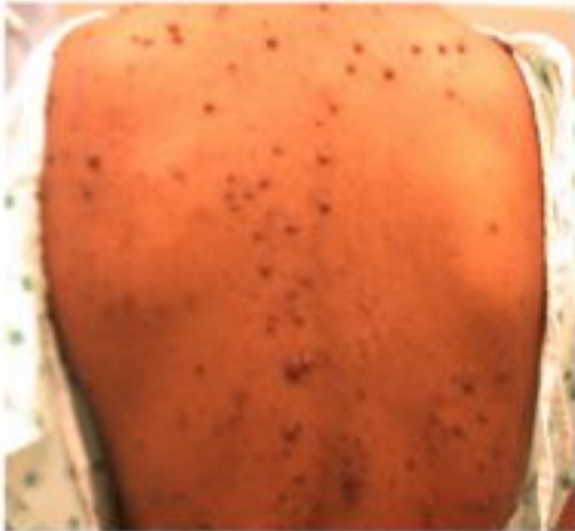
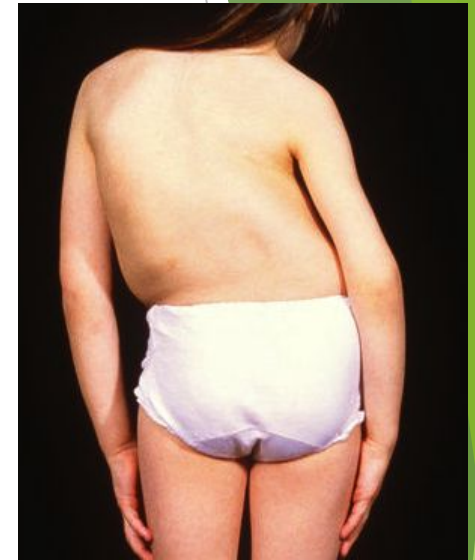
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Neurofibromatosis type 1 (NF1)

- ▶ Autosomal dominant condition affecting 1:3500
- ▶ Symptoms highly variable
 - ▶ Plexiform Neurofibromas (PNs)
 - ▶ Dermal tumors
 - ▶ Scoliosis
 - ▶ Glomus tumors
 - ▶ Chronic headaches
 - ▶ Gastrointestinal problems



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 - ▶ **Learning Disabilities/ADHD**



Pain in NF1

- Pain has been reported in 53% of youth with NF1 and PNs (Kim et al., 2009)
- Less information about pain in adults with NF1
- Our previous pilot study (n=12) provided preliminary support for ACT's feasibility and efficacy in adolescents with NF1 and chronic pain
- No randomized controlled trials of psychological interventions for people with NF1 and PNs who have chronic pain

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Online ACT interventions



- Trompetter, Bohlmeijer, Veehof & Schreurs (2014)
 - Randomized study of 238 adults with chronic pain
 - Pain interference: iACT group > control (and > WL when adherence was taken into account) at 3 months
- Buhrman et al., 2013
 - Randomized study: 7 weeks of ACT online readings and assignments w/ feedback from therapist
 - Acceptance (CPAQ): iACT group > online forum
- Simister et al., 2018
 - Randomized study of 67 adults with FM
 - iACT + TAU group improved on FM impact > TAU at 7 weeks



Results suggest that online ACT interventions are feasible and effective with adults with chronic pain.

Will it work for NF1?

NIH Study



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Primary Objective

- ▶ To evaluate the effects of a mixed-methods ACT intervention on pain interference among adolescents and adults with NF1, PNs, and chronic pain compared to a wait list control group

Secondary Objectives

- ▶ To evaluate effects of the intervention on pain intensity, pain anxiety, depression, quality of life, pain inflexibility, and pain acceptance

Methods

Eligibility

- ▶ 16-59 years of age with confirmed diagnosis of NF1 and a plexiform tumor
- ▶ Chronic pain for ≥ 3 months
- ▶ Pain interferes with functioning (PII mean score ≥ 2)
- ▶ Regular access to internet
- ▶ No new treatments for pain or tumors planned

Recruitment

- ▶ NIH cohort and NF registry

Intervention

In Person

- Two 2-hour sessions
- Patients learned and practiced mindfulness and defusion techniques
- Coping techniques re: unpleasant thoughts about their pain
- Completed workbook with values exercises and goal-setting

At home (8 weeks)

- Weekly email assignments
- Biweekly video chat
- Audio recordings of exercises

(Adapted from Dahl et al., 2005; Wicksell et al., 2007; Vowles et al., 2009; Masuda et al., 2011)

Measures

Domain	Measure
Pain Interference	Pain Interference Index (PII)
Pain Intensity	Numeric Rating Scale (NRS-11)
Pain Anxiety	Pain Anxiety Symptoms Scale (PASS-20)
Depression	Center for Epidemiological Studies of Depression (CES-D)
Quality of Life	PedsQL - NF Module (ADLs, Physical Fx)
Pain Acceptance	Chronic Pain Acceptance Questionnaire (CPAQ)
Pain Inflexibility	Psychological Inflexibility in Pain Scale (PIPS)

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Pain Interference Index (PII)

Pain Interference Index

Below you will find a list of questions about you and your pain. Please answer each question by circling a number between 0 and 6.

⊕ Please note that we are asking about your pain during the last week.

Has your pain:		Not at all			Some			Completely
1.	made it difficult for you to do work (in or outside the home)?	0	1	2	3	4	5	6
2.	made it difficult for you to do activities outside of work (leisure activities)?	0	1	2	3	4	5	6
3.	made it difficult for you to spend time with friends and family members?	0	1	2	3	4	5	6
4.	affected your mood?	0	1	2	3	4	5	6
5.	affected your ability to do physical activities (like run, walk <u>up stairs</u> , play sports, do chores)?	0	1	2	3	4	5	6
6.	affected your sleep?	0	1	2	3	4	5	6

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Statistical Plan

- ▶ Repeated measures ANOVA to look at group differences over time
- ▶ Paired sample T-tests to compare means within each group
 - ▶ Pool ACT and WL groups from pre to post tx
- ▶ 41 patients per arm will provide 80% power to test the differences in changes from baseline between the two arms
- ▶ **Target Accrual = 82**

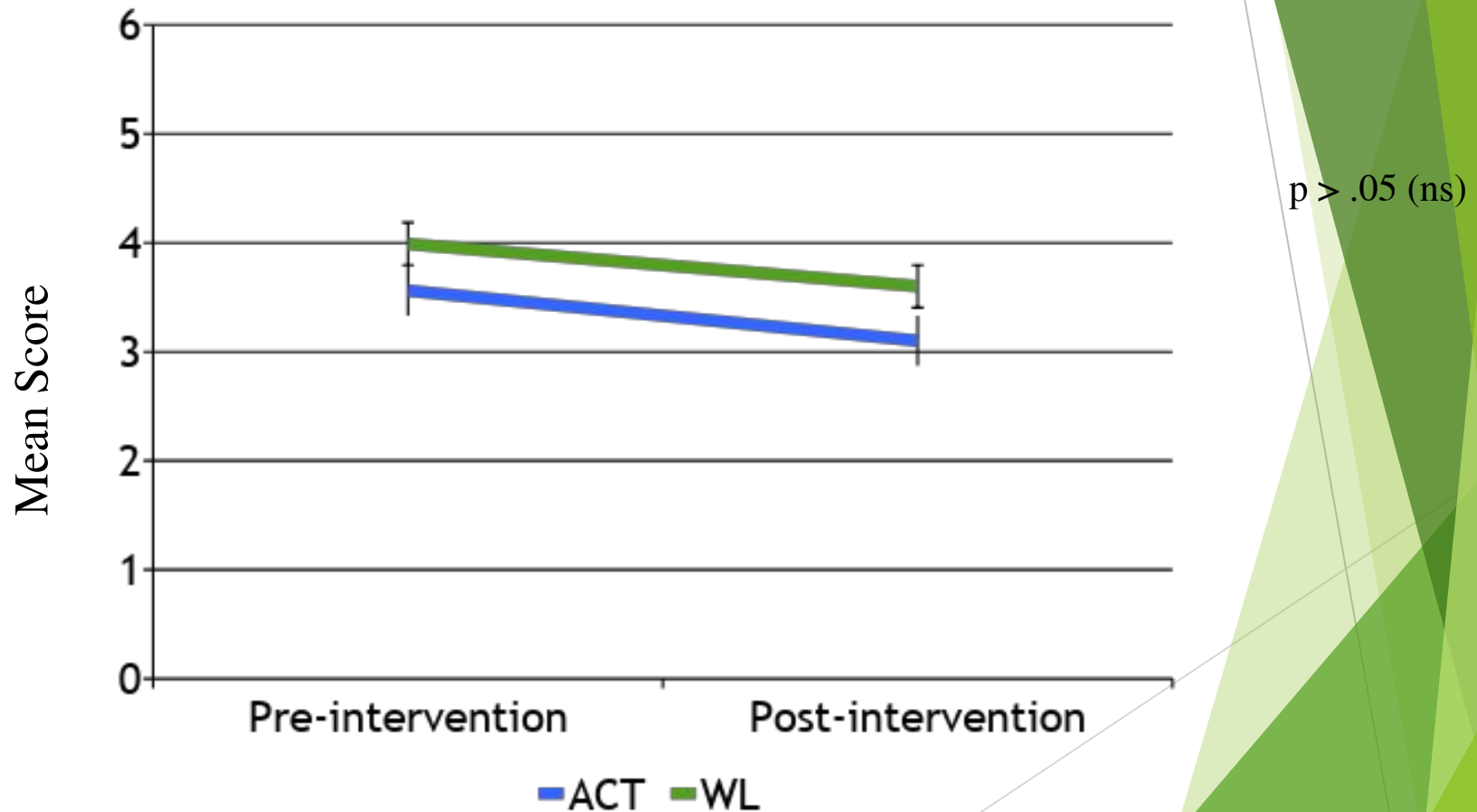
Results

Participants

- ▶ 50 participants enrolled
 - 1 (too many pain meds, could not do intervention)
 - 1 (genetic testing revealed did not have NF1)
 - 1 (too busy to complete intervention)
 - 1 (malignancy, progressive disease)
- 46 = Total sample in these analyses**
- ▶ Age: mean = 28.49 years, median 24.84 (range 16-56 yrs)
- ▶ Gender: 21 males, 25 females
- ▶ Race: 74% white, 11% black, 2% Asian, 13% mixed/unknown
- ▶ Ethnicity: 6% Hispanic

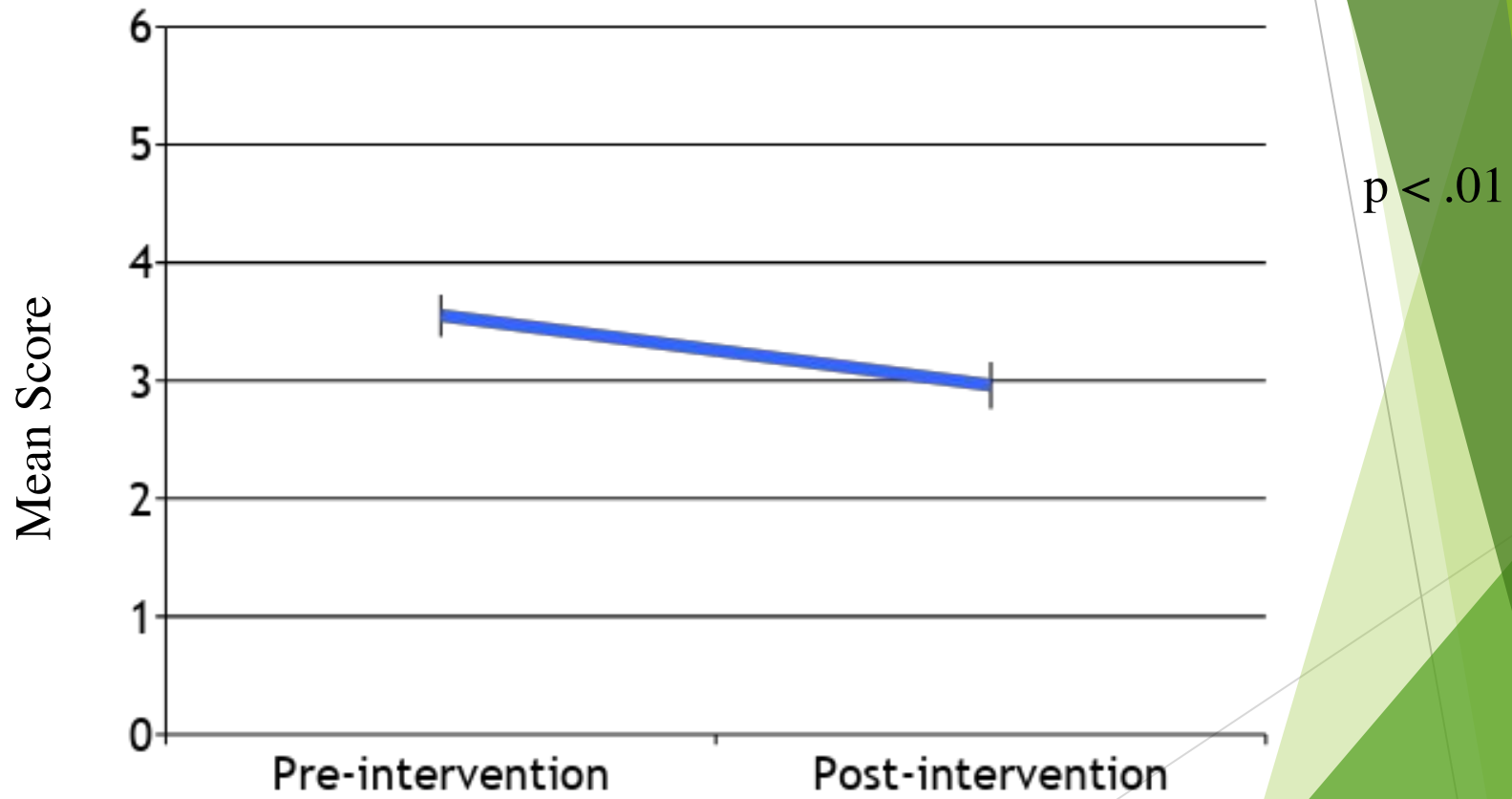
Results: Repeated Measures ANOVA

Pain Interference



Results: T-tests (ACT/WL pooled)

Pain Interference



Repeated Measures ANOVA

Secondary Outcomes

Variable	Mean (SD)				F
	Baseline	Post-tx	Baseline	Post-tx	
	ACT		Wait List		
Pain Intensity	7.5 (2.0)	6.6 (2.8)	7.1 (1.9)	6.6 (2.7)	.30
Depression	22.7 (12.5)	18.3 (9.6)	24.7 (13.2)	20.9 (11.4)	.073
Pain Anxiety	52.1 (21.5)	38.5 (20.6)	52.4 (23.3)	43.7 (20.5)	.96
QOL - Physical Functioning	42.8 (21.6)	52.7 (27.8)	42.5 (21.8)	43.8 (21.1)	2.03
QOL - Daily Activities	84.3 (12.8)	87.8 (10.6)	79.1 (23.1)	81.9 (16.2)	.02

All ps > .05

Repeated Measures ANOVA

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Repeated Measures ANOVA

Process Outcomes

Variable	Mean (SD)				F
	Baseline	Post-tx	Baseline	Post-tx	
	ACT		Wait List		
Acceptance	59.5 (15.1)	72.2 (15.1)	56.6 (14.1)	58.9 (15.8)	10.2
Pain Inflexibility	44.9 (13.5)	38.1 (14.2)	50.3 (13.8)	46.5 (15.7)	.85

$p < .01$

What we Know

- ▶ Pain interference significantly decreases post-ACT treatment among adolescents and adults with NF1 and chronic pain
 - ▶ Can work in people with learning/attention problems
- ▶ ACT interventions combining in-person and online components are feasible and reduce patient and clinician burden
- ▶ Acceptance is a key process

What we are Still Learning

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- ▶ Will effectiveness be demonstrated compared to control group when power is adequate?

In general

- ▶ Which processes are most important in facilitating change (and in which groups)?
 - ▶ Acceptance - pain willingness, activities engagement
 - ▶ Psychological Flexibility - avoidance, cognitive fusion
- ▶ How much is generalizable to younger adolescents and what modifications should be made?
- ▶ How low can we go? (how much therapist-to-client contact is necessary)
- ▶ How long do effects last? (we need longer follow-ups)

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My values...



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